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INTRODUCTION

The first objective of this work is to evaluate the social, emotional, and behavioral quality of life (QOL) of children with NF and explore potential linkages between NF and QOL in these 3 domains. The second objective of this work is to evaluate parental distress, family functioning, and child rearing practices. It is hypothesized that the social and emotional status of children with NF plays an important role in predicting eventual functioning and the quality of life they obtain. To determine which medical and psychological variables are involved, a cross sectional study of peer relationships, emotional well-being, and family functioning is required. Peer relations play a central role in children's social and emotional development and are fundamental for the development of adequate social skills and for the emergence of a healthy self-concept. Although a growing body of empirical evidence has shown peer perceptions of social competence are predictive both of current adjustment and of future adaptations during adolescence and through adulthood, no data are available on the peer relationships of children with NF. Additionally, while several psychometrically sound measures of emotional well being are available, these have not been reported for children with NF. Finally, we could locate no studies focusing on parent distress, family functioning, or child rearing concerns. A high incidence of learning disabilities, ADHD, and behavioral problems have been reported for children with NF. Clinically, parents often voice concerns that their children have few friends, are immature, and are teased because of their appearance or clumsiness.

BODY

Task 1: Months 1-4 Identify and recruit children with NF-1 (30 in Cinti/30 in Houston).

We have been able to identify and recruit 60 children with NF-1 in Cinti and Houston. We had 4 families decline to participate across the 2 sites.

Task 2: Months 1-6 Train staff to collect classroom data.

Staff were hired in Houston and Cincinnati to collect classroom data. The workshops were held and staff were trained. School districts were contacted and gave permission for anticipated future work. Materials were prepared for school data collection. For Houston, we trained staff to collect classroom data but they subsequently decided not to continue to work on the project. New staff were hired and they were trained.

Task 3: Months 5-11 Contact schools, collect data, enter, and score data.

No report, awaiting HSRRB approval.

Task 4: Months 5-11 Complete evaluations of disease severity for children with NF-1.

No report, awaiting HSRRB approval.

Task 5: Months 11-15 Use classroom rosters to recruit case controls who are closest in date of birth, same race/gender. Go to homes of children with NF-1 and case controls . . .

Staff were hired in Houston and Cincinnati to collect data in the home. Workshops were held and staff were trained. Materials were photocopied and readied for data collection. Awaiting HSRRB approval.

KEY RESEARCH ACCOMPLISHMENTS:

The approved project called for eight positions at the Houston site. Dr. Moore was already involved as the PI at the Houston site and Dr. Slopis as a Co-Investigator. In addition, the nurse coordinator and secretary positions were already in place. Extensive interviews were conducted for the positions of Project Coordinator and for the three Research Assistants. The Project Coordinator position was critical because it required a doctoral level person with substantial research experience who would conduct the day to day operations of the project. Eventually, we hired a doctoral candidate in Education Psychology from the University of Houston, Sandy Long. Ms. Long had previous experience as a licensed teacher and educational diagnostician as well as the research background we were seeking.

Considerable effort went into recruiting the research assistants in Houston. Two graduate students from the University of Houston were hired in January, 2000. They trained extensively (see below) for the school and home visit portions of the project. These two research assistants subsequently resigned in May and June, 2000, despite having been given assurances that they could stay with the project for the duration. The reasons for the resignations were in one case that the person's spouse was accepted to a training program in another state. In the other case the individual needed full time employment. Consequently, we were forced to begin the recruitment process again. Three new research assistants were hired over the period of late May to early August, 2000. These individuals are Amelia Molina, 5/29/00 to present; Bertha Sanchez, 6/19/00 to present; Vanessa Campbell, 8/7/00 to present. Staff have been trained in Houston to collect data in classrooms and homes. We have successfully completed this training with 4 staff on two occasions (2 staff each occasion).

Two protocols were drafted and submitted to the Psychosocial, Behavioral and Health Services Research Committee and subsequently to the Surveillance Committee of The University of Texas M.D. Anderson Cancer Center. These protocols correspond

to the school and home visit portions of this research project. Protocol P99-172 entitled "The Nature of Children's Peer Relationships" was approved and activated on August 3, 1999. The second protocol, P00-67, entitled "Psychosocial Functioning of Children with Chronic Illness and Their Families" was approved and activated on May 12, 2000. The protocols were initially approved in Cincinnati in 1989 for school based data collection and 1990 for the home based data collection, and have been annually renewed for the past 11 years. In Cincinnati, since the initial approvals we have collected data from approximately 25,000 children using the school based protocol, and approximately 600 children using the home based protocol (studying children with cancer, sickle cell diseases, juvenile rheumatoid arthritis, juvenile migraines, obesity, hemophilia, pilot study of children with NF, and children with mothers recently diagnosed with breast cancer). In Cincinnati, the school based protocol was last reviewed on 2/24/2000 and is approved until 2/24/2001. In Cincinnati, the home based protocol was last reviewed on 7/11/2000 and is approved until 7/11/2001.

Because the Houston group lacked the experience of the Cincinnati group in conducting studies such as this one, three training trips were conducted during the first half of 2000. The Research Coordinator (Long) and first two research assistants traveled to Cincinnati in late January. During this time they met with the Cincinnati personnel for two days to learn the methods and procedures to be used in the school visit portion of the project. This included interview techniques to successfully recruit school principals and teachers, ways to ensure subject confidentiality, and the actual administration of the different study measures. Role playing was done to help the trainees understand possible scenarios they might encounter in the classrooms or in the interviews with teachers and principals. Training was also accomplished in the methods of data collection, the importance of maintaining confidentiality, data entry, and data scoring. These procedures have been used by the Cincinnati group for 10+ years and are very detailed. Because of the complexity of data management procedures, we felt it was necessary for one of the Cincinnati personnel to travel to Houston to give further training. This was done in late March, 2000.

Because of the turnover in personnel described, it was necessary to repeat training for the three new research assistants. The Houston Research coordinator (Long) accomplished much of this training, but we also felt it was necessary to have training by the Cincinnati group. For this reason, Dr. Noll (Cincinnati PI) traveled to Houston in late July for a two day training session with the new personnel. This included training not only on the methods and procedures for the school visit portion of the project, but also the home visits.

Some of the procedures of the project are very sophisticated and require a considerable amount of training and practice. For this reason, the research assistants and the Project Coordinator have been practicing these procedures on each other and

on healthy volunteers who come to our clinic with families of patients. This training has gone very well and all personnel are now highly competent in these procedures.

This project is very labor intensive. Materials must be prepared for each school principal and teacher to explain the project and procedures. These packets were assembled by the research assistants with the assistance of the secretary on the project. In addition, there is an enormous amount of photocopying required. We anticipated recruiting 30 subjects at each site during the first year. We expected each subject to have 25 classmates. Each subject and classmate (expected to be 1500 at the two sites) would receive a number of items for completion in the classroom portion of the project resulting in an enormous amount of paperwork that must be accurately photocopied and assembled. The project secretary accomplished these tasks and also acquired the correct test booklets and forms necessary. These were all assembled into packets.

REPORTABLE OUTCOMES: Staff have been hired in Houston to manage the project (Ms. Long) and she has participated in the training of 4 research assistants. Training of staff in Cincinnati was also accomplished within the context of other funded research using this design and methodology. Materials were prepared at both sites for data collection.

CONCLUSIONS: It is feasible to identify appropriate staff and get them trained within the parameters we describe in the grant. A comprehensive training manual has been developed for future use. The institutional review boards at Children's Hospital Medical Center and MD Anderson both reviewed the protocol favorably. The HSRRB has not approved this research protocol.

REFERENCES: None

APPENDICES: None

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